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SHORT REPORT

Neck Mass Caused by Spontaneous Arteriovenous Fistula of the Superior Thyroid Artery to its Homonymic Accompanying Vein by Duplex Ultrasonography

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Abstract An arteriovenous (AV) fistula is an abnormal connection between an artery and vein. It is usually caused by congenital and acquired factors. A 65-year-old man presented to us with a neck mass of 4-year duration with no traumatic history. A Doppler study of the mass revealed a 'whirlpool' pattern in a cavity, and something resembling a thrombus adherent to the cavity wall. Based on the spectrum of blood flow, we strongly suspected a fistula. Digital subtraction angiography (DSA) was carried out to validate that the neck mass was caused by an AV fistula from the superior thyroid artery (STA) to the superior thyroid vein (STV).

Arteriovenous (AV) fistulae of the superior thyroid artery (STA) are rare (particularly those that occur spontaneously). An AV fistula often appears as a pulsatile mass. This mass can thrill upon palpation, cause a continuous murmur through auscultation and may be accompanied by higher skin temperature.

The examination of a pulsatile mass should initially be done by palpation. The perception of palpation for a pulsatile mass depends on the velocity of blood flow within the fistula and changes in the corresponding vein. Most AV fistulae, in general, are caused by trauma or by medical procedures.[1,2] Doppler ultrasonography is the most convenient and cost-effective procedure for detecting AV fistulae: it can accurately detect the location of the fistula and the vessels involved.

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Case report

A 65-year-old Chinese man who had been taking antihypertensive medication for the previous 10 years presented to us with a small mass originating at the right side of the neck. It had been growing slowly without inflammatory changes such as redness, swelling, increased temperature or pain. He had no history of trauma.

Physical examination showed that the mass was located on the right side of the neck just above the right thyroid, $\sim 3 \times 3 \times 5$ cm in size with a moderate texture, an ill-defined boundary and limited mobility, and caused no pain under pressure. A localised vascular murmur was audible. The thyroid was normal upon palpation. Blood counts, routine blood chemistry and serology were negative or within the normal range.

Gray-scale ultrasonographic examination showed that the mass was an irregular and tortuous cyst-like area above the right lobe of the thyroid gland (but with no relationship to the thyroid gland), extending to the submental triangle. The mass was anechoic and matter resembling a thrombus was at the periphery (Fig. 1a). Turbulent flow was detected in the cavity by Doppler imaging (Fig. 1b). At the submental triangle, rapid blood flow was detected during the entire cardiac cycle. The highest speed was ~ 300 cm/s and was displayed by pulsed-wave Doppler (PWD; Fig. 1c). While the Doppler sampling volume was set in a lower position of the cyst-like area, the spectral form was characterised by arterialised vein spectral form (Fig. 1d) or low-resistance artery spectrum. This indicated that the mass should be an AV fistula as far as the spectral changes were concerned. Then we identified the common artery (CCA) and the internal jugular vein (IJV) on both sides. A significant difference between the CCA and IJV was not observed with respect to the spectrum. Entry of any feeding artery and the draining vein was not identified by ultrasound.

Angiography demonstrated enlarged arteries and veins, with arterial supply typically derived from the right superior thyroid artery (Fig. 2). The vein was the homonymic

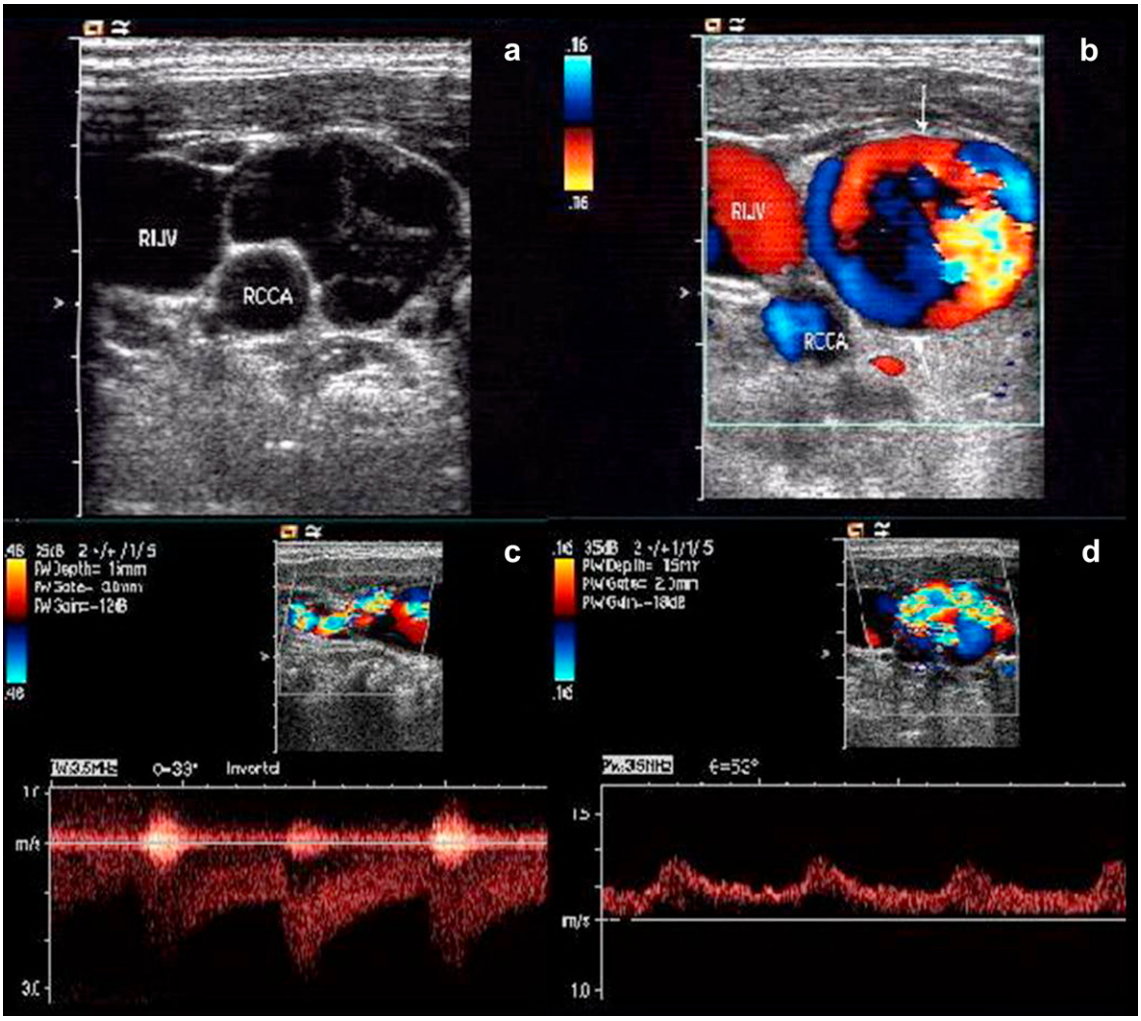

Figure 1 The right-side neck mass was anechoic; a thrombus was in the periphery (a). Doppler ultrasonography showed turbulent blood flow in the cavity (b). High-speed blood flow during the entire cardiac cycle, the highest speed was about 300 cm/s as displayed by PWD (c). Arterialised spectrum (d) was observed in the dilated vessels. RCCA: right common carotid artery; RIJV: right internal jugular vein.

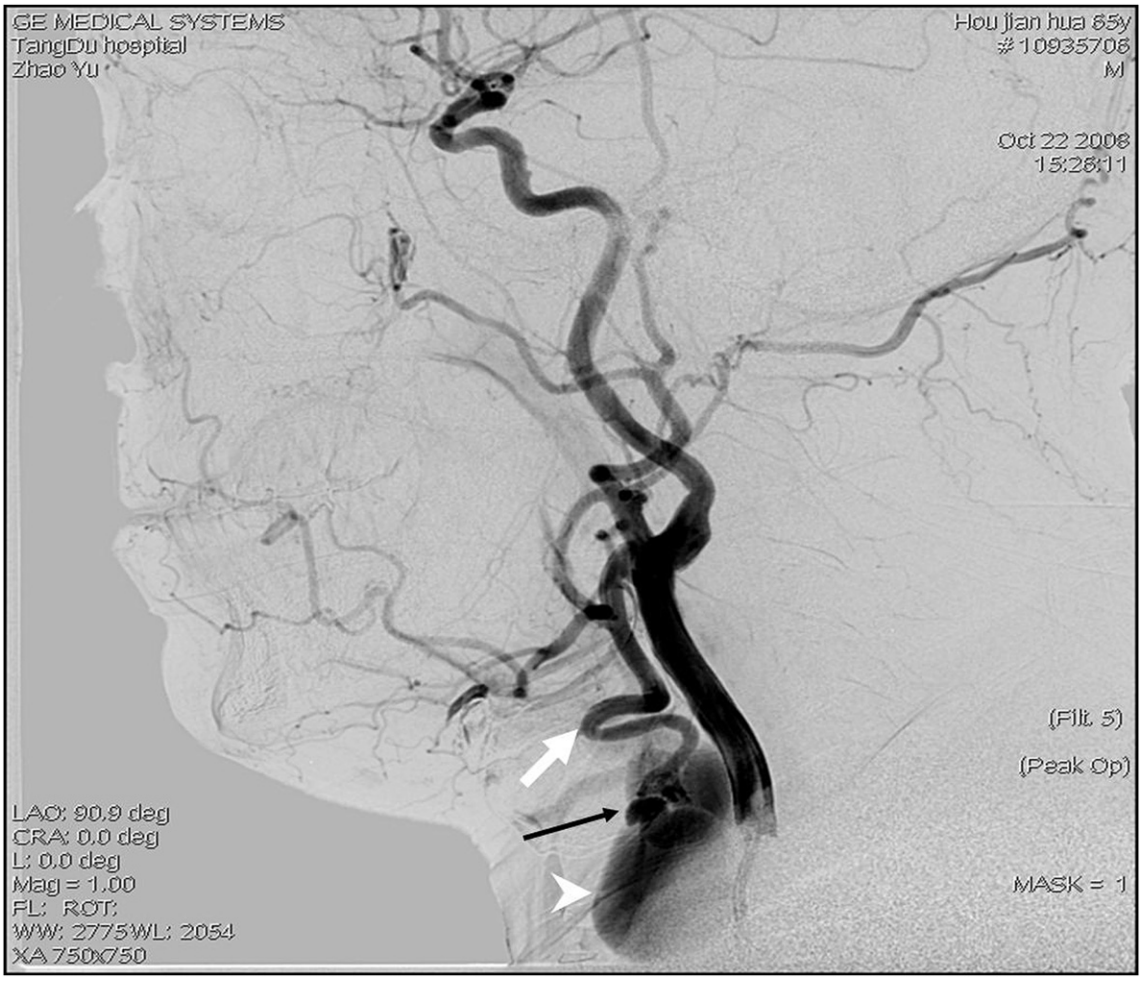

Figure 2 DSA showed that there was a shunt (black arrow) from the right tortuous STA (white arrow) to the adjacent dialated vein (white arrow head), with the latter flowing into the right IJV.

accompanying vein. The patient underwent selective catheterisation of the STA followed by embolisation with 34-Onyx (Micro Therapeutics, Inc, Irvine, CA, USA). Post-embolisation arteriography revealed occlusion of the feeding vessel and obliteration of the abnormal collection of vessels.

Discussion

Aneurysms and AV fistulae can cause neck masses that are pulsatile in appearance and appear hyper-vascular on colour-Doppler imaging. An AV fistula can cause dilation of both supplying artery and draining vein. Some AV fistulae,

Table 1 Main parameters of the differential diagnosis between aneurysms and fistulae.

Parameters	True aneurysm	False aneurysm	AV fistulae
Cause	Smoking and hypertension	Trauma or iatrogeny	Trauma or iatrogeny
Physical examination	Pulsatile mass		
Gray-scale imaging	Complete artery wall; focal dilated lumon; mural thrombus	Incomplete artery wall; surrounding soft tissues; mural thrombus	Incomplete wall between artery and vein; dilated involved vessels; mural thrombus in draining vein
Colour-Doppler	'Whirling' flow in the aneurysm	Bright systolic blood flow from the breakage	Bright blood from the breakage in the whole cardiac cycle
Spectral-Doppler	Little change	''Out with in'' form	''Out without in'' form

especially those located superficially as a localised form, have a sponge-like appearance. In the vein, there is a 'whirling' flow pattern in the dilated lumen on colour-Doppler imaging. The artery also becomes wider owing to lowered resistance and accelerated circulation from the shunts. Spectral analysis shows the change in resistance of the feeding artery and arterialised spectrum of the draining vein as the reason for arterial pressure transmitting directly to it.[3] A mural thrombus in the distal end of the draining vein may be observed. AV fistulae are usually caused by trauma or have an iatrogenic cause.

One must distinguish between aneurysms and AV fistulae. A pseudo-aneurysm formation is the most common complication resulting from arterial catheterisation.[4] The aneurysms are contained by surrounding soft tissues, which form a thin fibrous capsule rather than the true layers of the arterial wall. Trauma is the other main non-iatrogenic cause. A true aneurysm has a confined lumen, which is defined as focal dilatations at least 1.5-times the diameter of the adjacent normal artery. Arterial aneurysms are usually seen in male patients older than 70 years[5]; smoking and hypertension are other risk factors.

Spectral analysis is a very important parameter to differentiate the AV fistula and pseudo-aneurysm. In pseudo-aneurysm, high-speed systolic blood flow from the artery to the outside and low-speed diastolic blood flow from the outside to the artery is shown, resembling an 'out-with-in' spectral form. While, in AV fistula, if the fistula can be traced, the spectrum at the fistula site is characterised as an 'out-without-in' blood spectrum — that is, only the high-speed systolic blood flow from the supplying artery to the draining vein can be detected. The important parameter for the differential diagnosis is listed in Table 1.

The STA is an invariable feeding artery for the thyroid. It rises from the anterior surface of the external carotid artery (ECA). We can readily detect the origin of the STA by ultrasound, but only ~1 cm of its length can be traced. There are few reports about the clarity of visualisation using Doppler ultrasonography for the distal STA. It is usually quite difficult to detect the STA by ultrasound, particularly, if a patient has a short, thick neck or a higher bifurcation. In addition, the feeding artery and draining vein become tortuous, thereby obscuring the images. A shunt occurring at the distal STA is hard to detect using ultrasound. We considered the mass to be an AV fistula because of the spectrum we obtained. Our primary diagnosis was validated by digital subtraction angiography (DSA) and also the involved artery and vein.

Ultrasound can usually document the vascular abnormalities by colour-Doppler imaging and the spectrum if the image is sufficiently clear. In some cases, the spectral form is more important for the differential diagnosis. If the image is not qualified by Doppler ultrasound, DSA or magnetic resonance angiography (MRA) is used, which has a very important role in the diagnosis of AV fistulae for mapping out the characteristics of AV-malformation flow. It is also used for treating AV fistulae. DSA is invasive and more expensive than ultrasonography; so the latter is the ideal imaging technique for rapidly establishing the correct diagnosis.

AV fistulae of the neck are well-known problems of venous puncture of the internal jugular vein. They are predominantly known to occur between the carotid artery and the internal jugular vein.[6] It is in the range of possibilities that it happens also between smaller vessels in this anatomic region, for example, the STA and the superior thyroid vein (STV). Spontaneous AV fistulae of the STA and STV are very rare, and the pathogenesis is not exactly understood. There is one report about renal fistulae caused by hypertension.[7] Our patient had a 10-year history of hypertension, which provided a clue to the aetiology.

Conflict of interest/funding

None.

Ethical approval

The article has received the ethical approval for research.

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